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Myofibroblastoma of the breast: Case report and literature review

Marco Mele[a,*], Vibeke Jensen[b], Adam Wronecki[c], Giedrius Lelkaitis[d]

[a] Surgical Dept., Aarhus University Hospital, Denmark
[b] Pathology Dept., Aarhus University Hospital, Denmark
[c] Radiology Dept., Aalborg University Hospital, Denmark
[d] Dept. of Pathology, Aalborg University Hospital, Denmark

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ABSTRACT

Myofibroblastoma of the breast is a rare benign mesenchymal tumor. The literature describes relatively few cases of this type of tumor. We report on a new case of myofibroblastoma in a 65-year old man successfully managed at our institution. The purpose of this case report is to highlight characteristics and differential diagnosis of this rare neoplasm.

1. Introduction

Myofibroblastoma of the breast is an uncommon benign stromal tumor, predominantly occurring in menopausal women and older men. The clinical presentation is characterized by a mobile, well defined and solid palpable tumor. Histologically myofibroblastoma is well demarcated and composed of bipolar spindle cells structured collagen bundles. Based on histological, immunohistochemical and ultrastructural observations, tumor cells have mesenchymal origin and show myofibroblastic differentiation. This type of tumor causes differential diagnostic problems by mammography and ultrasonography, as it appears in several different variations and for example can be confused with hamartoma or fibroadenoma.

2. Case presentation

A 65-year-old man was referred to our institution for a large lump in the right mammary region. He was an otherwise healthy man, although on medications for hypercholesterolemia and hypertension. There was no family history of breast cancer. The lump, which initially appeared as a small and asymptomatic swelling, had slowly enlarged for more than a decade. On examination, there was a large tumor located underneath the areola and measuring 13 cm of maximum diameter (Fig. 1). The lesion was multilobulated, well-defined and freely mobile with respect to the underlying muscular plane, but adherent to the overlying skin. The tumor was clinically considered benign and there was no regional lymph nodes involvement. Mammography and ultrasonography showed a smooth well-defined tumor measuring 10 cm × 8 cm (Fig. 2). Ultrasonography also displayed a mixed echogenic gland and adipose tissue. Tru-cut biopsy was suggestive of adenomyoepithelioma or pleomorfic adenoma. Therefore, the tumor was completely excised with the overlaying skin and preserving the nipple (Fig. 3). The post-operative period was uneventful. Gross examination revealed a 10-cm, well-demarcated nodular tumor with reddish brown cut surface and finely lobulated adipose tissue in the central area. There was a free resection margin measuring 5–10 mm.

Tumor tissue blocks were fixated in neutral buffered 10% formalin for 48 h. 3–5 μm slides were made from paraffin embedded tissue blocks and stained routinely with haematoxylin–eosin (HE). Immunohistochemistry was performed using DAKO Autostainer Universal Staining System (Figs. 4 and 5).

Histological examination revealed a well-defined and pseudoencapsulated tumor, surrounded by adipose tissue. Tumor was cellular and vaguely nodular, with mesenchymal appearance, devoid of breast parenchyma and with abundant "entrapped" islands of adipose tissue or separate adipocytes. Tumor cells were bipolar, monomorphic, showing oval nuclei without atypia and with very scant mitotic activity. Cells were arranged in ill-defined fascicles, haphazardly intermingled with coarse short bundles of collagen. There were no areas of necrosis, hemorrhage or metaplastic changes. Immunohistochemistry revealed a positive reaction of tumor cells for actin, desmin, vimentin, caldesmon and CD34, while reaction for S100 and cytokeratin 7/19 was negative. The proliferative fraction of tumor cells, detected with Ki-67, was 1–5%. Present findings were consistent with a diagnosis of myofibroblastoma.

3. Discussion

Myofibroblastoma was first characterized by Wagortz et al. in 1987, reporting on 16 of such cases.[7] This tumor is a rare and benign mesenchymal lesion, which shares similarities with other

* Corresponding author. Tel.: +45 89499674.
E-mail address: mele_marco@hotmail.com (M. Mele).

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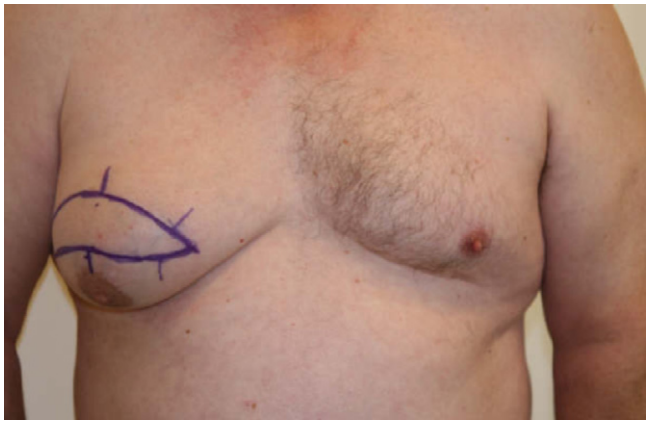

Fig. 1. Asymmetry with great palpable mass in the right side.

tumors as spindle cell lipoma-like tumor, solitary fibrous tumor, myofibroblastoma, leiomyoma and benign fibrous histiocytoma. All these lesions are classified as benign spindle cell neoplasms, which may show fibroblastic, myofibroblastic and fibrohistiocytic aspects.[2,5,6,10,12,17] Perhaps, they all derive from the same type of a mesenchymal cell precursor and exhibit a spectrum of overlapping histological and immunophenotypic properties, where differentiation in myofibroblastic direction, among others, is characterized by positivity for antibodies against α-smooth muscle actin and desmin.[4] Reported cases of myofibroblastoma occurs most often in women and men aged 40–87 years.[8] There is a tendency that myofibroblastoma has a penchant for older men and postmenopausal women. There are no reported cases that indicate relation to gender, race, medical conditions, use of medication or other effects of growth factors. It is crucial to understand the molecular mechanism, controlling growth and proliferation of myofibroblasts. It is believed that this mechanism is somewhat related to autocrine and paracrine secretion of cytokines, which transform growth factors in a particular way (TGFβ), but it is also demonstrated that specific tumor necrosis factor (TNF) and fibroblastic peptide-trophic growth factors are generally present in these tumors.[3] Differential diagnostic considerations include a number of mesenchymal/soft tissue lesions and neoplasms, and fibromatosis, which, unlike myofibroblastoma, is characterized by infiltrative growth and is negative for CD34.[9] Inflammatory myofibrolastic tumor is characterized by spindle shape myofibroblastic cells with accompanying infiltration of lymphocytes, plasma cells and histiocytes, which is not found in this case.

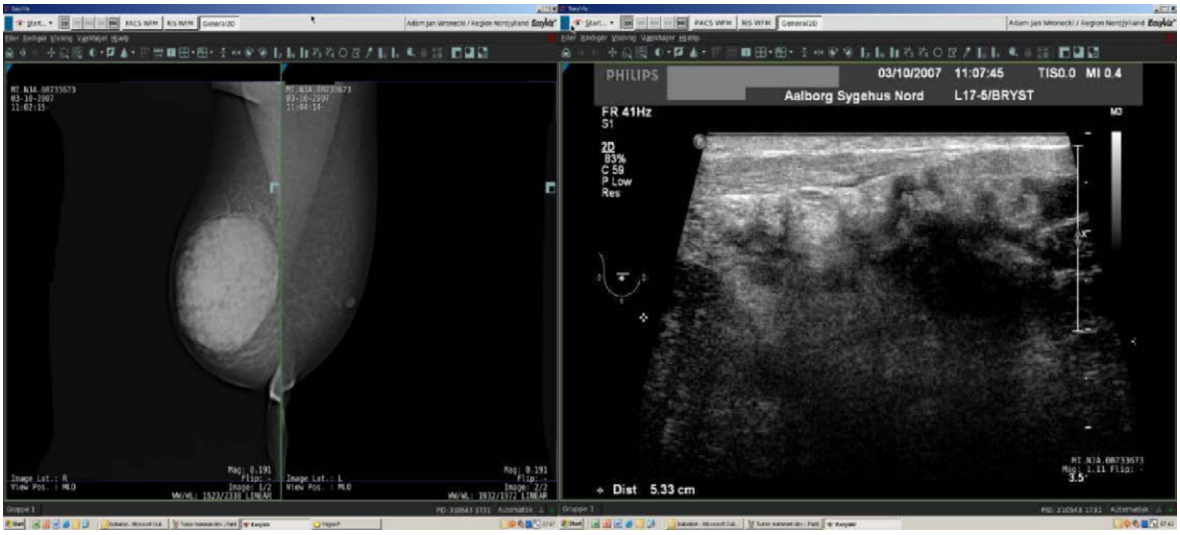

Fig. 2. Typical images of myofibroblastoma from mammography and ultrasound.

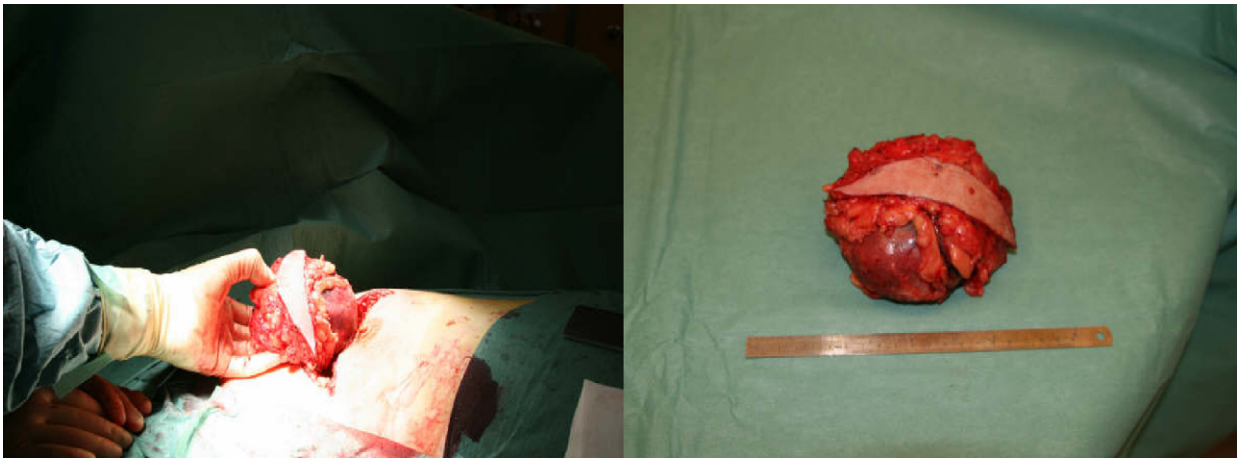

Fig. 3. Images from the operation and the specimen.

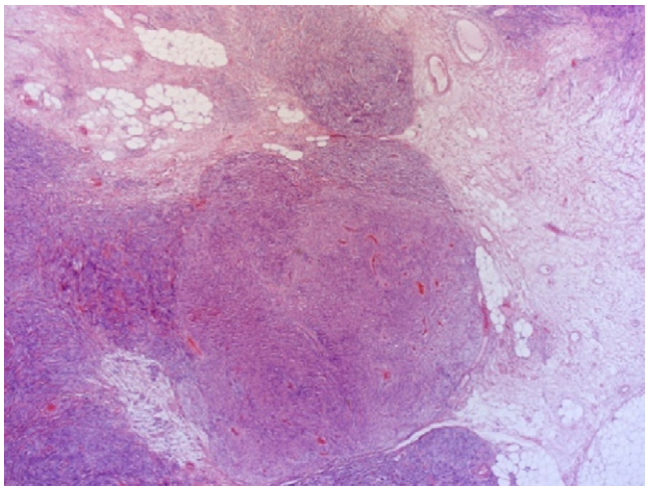

Fig. 4. HE, 20×. Well-defined nodular tumor with "entrapped" islands of adipose tissue.

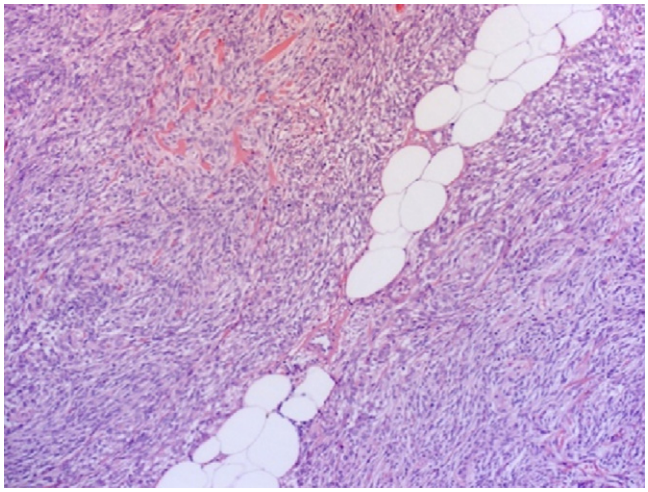

Fig. 5. HE, 100×. Fascicular bundles of bipolar tumor cells with few collagen fibers and areas of adipose tissue.

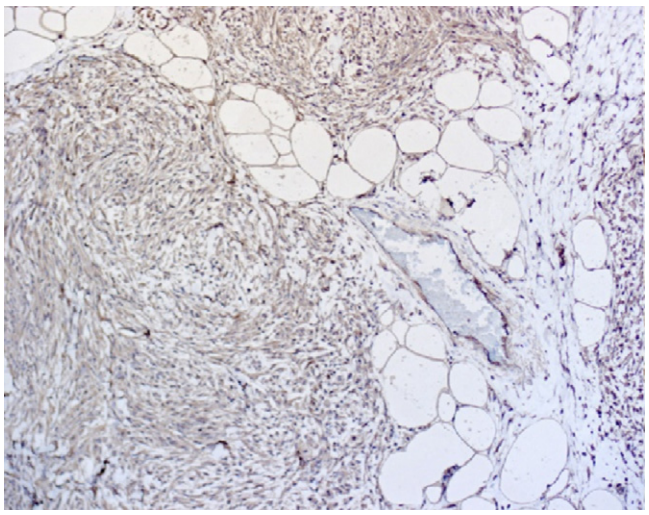

Fig. 6. ASM-Actin, 100×. Tumor cells with positive reaction, positive internal control (smooth muscle cells in vascular wall).

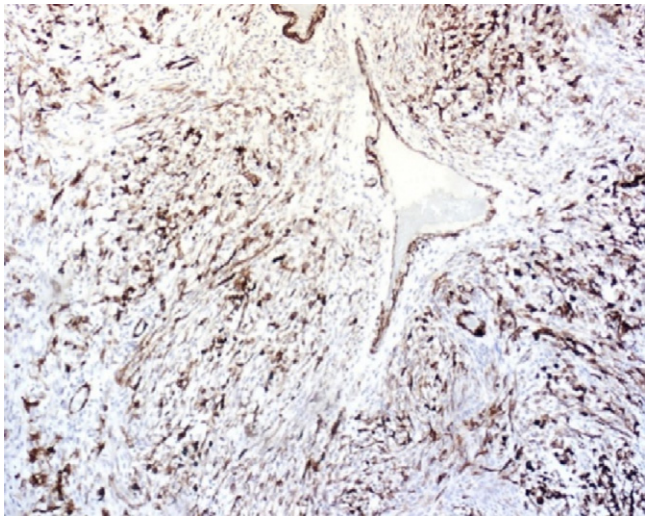

Fig. 7. h-Caldesmon, 100×. Focal positivity in tumor cells.

The tumor is negative for S-100, which is typically positive at Schwannom and spindle cell lipoma. Morphology of tumor cells and immunohistochemical positivity for α-smooth muscle actin, desmin, vimentin and CD34 are in line with myofibroblastic differentiation of the tumor. Whereas positivity for h-Caldesmon suggest leiomyomatosis differentiation (Figs. 6–9).[18] The malignant variants of spindle cell presents typically greater cell density, marked nuclear pleomorphism, high mitotic activity with atypical mitotic figures. It has been extensively debated whether solitary fibrous tumor and myofibroblastoma of the breast should be considered as two distinct lesions. Indeed, there are some recent reports considering these rare lesions as histologically different.[11] Myofibroblastoma is most often described in women, who usually present with an asymptomatic and slowly enlarging breast tumor. Mammography typically shows a heterogeneous well-defined encapsulated tumor without microcalcifications. Ultrasonography usually demonstrates a well demarcated tumor, although a variable and mixed echo pattern can be expected, sometimes with more distal acoustic attenuation as a result of incorporation of fat tissue and other types of tissue in tumor. Doppler modality may show a slight peripheral hypervascularization of the tumor. Accord-

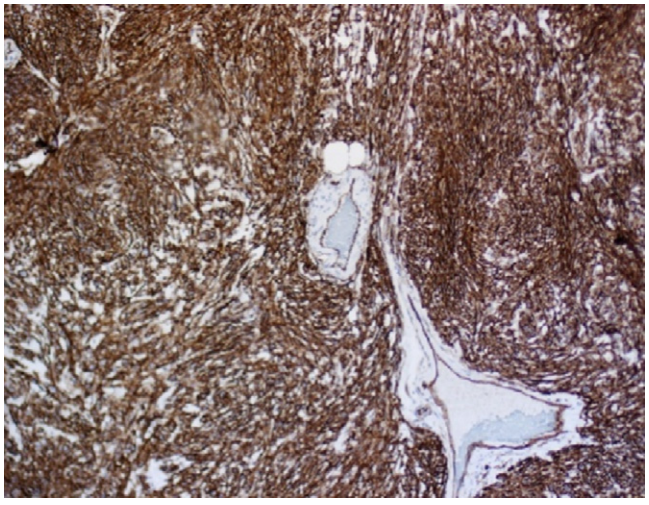

Fig. 8. CD34, 100×. Strong positive reaction in tumor cells, positive internal control (endothelial cells).

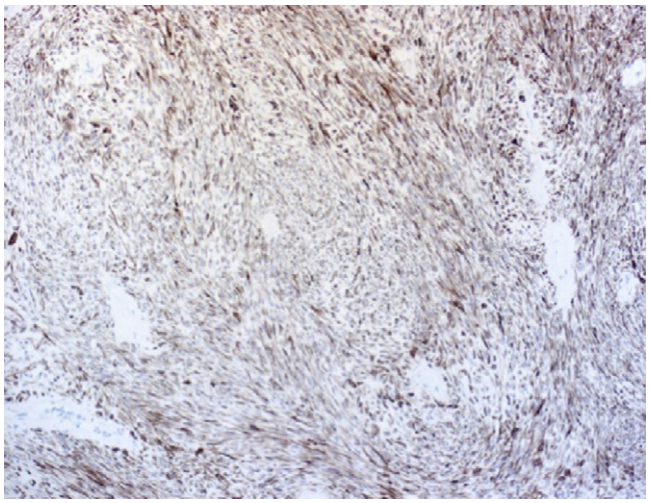

Fig. 9. Desmin, 100×. Generally positive reaction in tumor cells.

ing to the imaging, the differential diagnosis includes hamartoma, fibroadenoma and lipoma. Regarding the tumor size, most of the reported cases dealt with lesions measuring from 1 to 3.7 cm of maximum diameter. However, the tumor may attain very large dimensions as present case and others recently published.[1] We concur that Tru-cut biopsy is a reliable procedure in order to obtain histological diagnosis before planning complete surgical excision of the lesion.[13–16]

4. Conclusion

Myofibroblastoma is a well capsulated tumor with a good cleavage plane, which usually allows easy surgical excision. Surgery is the recommended treatment and, as long as the resection margins are free, relapse is unlikely. Additionally, malignant transformation has not been reported yet. However, a minimum of 24 months' follow-up is desirable.

Conflicts of interest statement

None

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None

Ethical approval statement

We hereby state that written consent has been obtained from the patient and will be provided upon request.

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